

CASE REPORT

Multiple basal cell carcinomas following x-ray treatment of tinea capitis in childhood: a case report and a literature review

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Summary

Introduction: Basal cell carcinoma (BCC) is the most frequent skin cancer with a rising incidence worldwide. Predilection sites are the head and the neck in 80-85% of cases. Leading risk factors are sun exposure and ionizing radiation. In the middle of the 20th century, radiation-induced epilation was used as an efficient treatment method for tinea capitis.

Patient review: We report a case of an 80-year-old man with a 16-year history of the successive occurrence of multiple BCCs on the scalp, face, and neck. He also had the history of tinea capitis treated with radiotherapy during childhood. At presentation during clinical and dermoscopic examination approximately 25 BCCs were observed, predominantly on the scalp. The most common BCC lesion was the nodular subtype, followed by the pigmented and superficial subtypes. Histopathological examination confirmed the diagnosis of BCC in multiple lesions and one squamous cell carcinoma (SCC). In personal history, the most important comorbidity was hairy cell leukemia. The therapeutic approach included surgery and 5-fluorouracil 5% cream.

Conclusion: Radiation-induced epilation was used as an efficient method for treating tinea capitis. One of its side effects was the appearance of BCC in radiation exposed areas. Surgical excision is the gold standard for BCC treatment. Also, 5-fluorouracil 5% cream may be a good option for patients with multiple BCCs. We present satisfactory results after combined treatment in our patient.

Key words: basal cell carcinoma, ionizing radiation, radiotherapy, tinea capitis, treatment

INTRODUCTION:

Basal cell carcinoma (BCC) is the most frequent skin cancer with a rising incidence worldwide. Most cases are located mainly on sun-exposed areas such as the head and the neck (80-85%) and are locally invasive, indolent and characterized by slow growth and rare metastases (1). In the middle of the 20th century, radiation-induced epilation was used as an efficient treatment for tinea capitis (2, 3). Radiotherapy of the scalp during childhood is a widely recognized risk factor for BCC (4) and it is the subject of numerous published studies (5-15). We present a patient with multiple BCCs, solitary squamous cell carcinoma (SCC) and hairy cell leukemia.



Figure 1. Multiple BCCs on the head and neck and SCC on the left auricle

CASE REPORT

We report a case of an 80-year-old male with a 16-year history of the successive occurrence of multiple BCCs on the



Figure 2. Dermoscopic findings: comma like vessels, haemorrhage, hypopigmented areas and telangiectasias.

scalp, face, and neck. On admission during clinical (**Figure 1a and Figure 1b**) and dermoscopic examination approximately twenty-five BCCs were observed. The most common type of BCC was the nodular type (**Figure 2**), followed by pigmented and superficial subtypes (**Figure 3a and Figure 3b**). On the left auricle there was one nodular lesion repetitively bleeding on gentle touch (**Figure 4**). In personal history he reported radiotherapy for scalp tinea at the age of 12, hairy cell leukemia at the age of 59, hypertension, arrhythmia and benign prostatic hyperplasia. Multiple BCCs started to appear at the age of 64, which is 52 years after radiation exposure. Hairy cell leukemia had been treated with purine analogue cladribine (2-chlorodeoxyadenosine) for 19 years until remission. Family history was negative for skin or other malignancies.

Routine laboratory tests, including complete blood count, biochemical analysis, tumor markers and urinalysis, were within normal limits. Histopathological examination confirmed the diagnosis of BCC in multiple lesions and SCC in a nodular lesion on the left auricle. Neck ultrasonography showed no pathological findings.

The treatment included surgical excision for multiple BCCs (over 15 times) and solitary SCC on the left auricle, and the topical treatment with 5-fluorouracil 5% cream for 6 weeks for inoperable BCCs (**Figure 5a and Figure 5b**). A great compliance with the patient was achieved.

DISCUSSION

BCC is the most common skin cancer with an increasing incidence worldwide (1). Treatment comprises surgical excision as the gold standard, topical therapy with 5-fluorouracil 5% or 5% imiquimod cream and sonic hedgehog inhibitors vismodegib and sonidegib for locally advanced

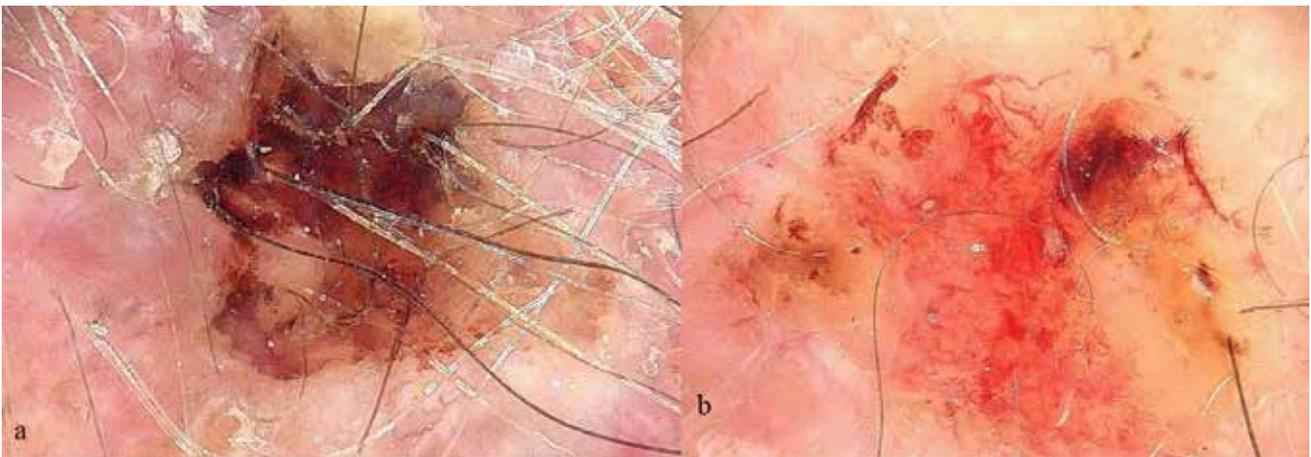


Figure 3. Dermoscopic findings: specks of brown and grey pigment, focal ulceration (left photo); milia, disordered and streaky crystalline structures and white rim around central ulceration (right photo).

and metastatic BCC (6). Exposure to UV radiation and previous ionizing radiation have been recognized as main risk factors for BCC in literature (5, 7, 8).



Figure 4. Multiple BCCs on the head and solitary SCC on the left auricle.

Albert et al. in 1968 (9) reported the first study of long term effects after x-ray treatment for tinea capitis in childhood. High prevalence of multiple BCCs was reported. The most common histological type was nodular, like in our patient. A similar study was published by Maalej et al. (10). In this study, BCCs and SCCs were the most common radiation-induced skin cancers, children were between 6 and 18 years of age and skin cancers started to appear 22 to 50 years after radiation exposure. In reviewed literature, there are data on an inverse relationship between the occurrence of BCC and age when the irradiation occurred. (8, 11). Boaventura et al. also found that there was a greater risk of developing BCC if the radiation exposure occurred at a younger age (12), which is similar to our patient who was irradiated in childhood.

Shore et al. reported comparative study in which 3,604 children participated and were followed up to determine the incidence of cancer after a follow-up period of up to 50 years. The children were divided into two groups, the first containing patients who were treated with X-ray therapy and the second containing those who were treated only with topical medications (13). In the irradiated group, about 40% of patients developed multiple BCCs and only 0.31% SCCs. This study also indicated the importance of skin phototype and sensitivity to UV radiation as well as that children irradiated at a young age had the highest BCC risk.

Mseddi et al. (14) reported 33 patients with diagnosed scalp BCC who were treated in childhood with radiation therapy due to tinea capitis. The median age at the moment of the irradiation treatment was 9 years. The latency period between the received X-ray therapy and the onset of BCC ranged between 21 and 51 years, and in our patient, the first lesion appeared after 52 years. Both clinically and histologically, nodular BCC was the predominant tumor subtype, which is the case in our patient as well.

Another retrospective study from Iran evaluated the difference between previously irradiated and nonirradiated group of patients with BCC tumors during a 10-year follow-up (15). Patients with a history of childhood therapeutic radiation had a more aggressive form of BCC, and needed more extensive surgical treatment, while the recurrence rate of BCC was significantly higher.

In the reviewed literature, BCC was described in the largest number of patients, as a result of radiation exposure of the head and neck. However, there are also data according to which patients developed more malignant diseases (10, 16), like in the case of our patient who developed three malignancies: BCC, SCC, and hairy cell leukemia. The previously mentioned study by Shore et al. also assessed the risk of tumors and malignancies following childhood irradiation treatment for ringworm of the scalp. They found an apparent excess of hematological malignancies in the irradiated group. Hairy cell leukemia was diagnosed in one patient (16).



Figure 5. Scars and residual pigmentation after combined treatment.

CONCLUSION

Radiation-induced epilation as a treatment option for tinea capitis showed to be associated with multiple side effect such as BCC and SCC development. Since sonic hedgehog inhibitors may be promoters of cutaneous SCC (17, 18), they were not considered as a therapeutic option in our patient. Nevertheless, we achieved satisfactory treatment results with occasional surgical excisions of

solitary lesions and topical treatment with 5-fluorouracil 5% for superficial BCCs.

Author contribution

All listed authors contributed equally to the conception of the work, the interpretation of data, preparing the draft of the manuscript and the interpretation of the revised version.

References

- Clio Dessinioti, Christina Antoniou, Andreas Katsambas, Alexander J Stratigos. Basal cell carcinoma: what's new under the sun. *Photochem Photobiol.* 2010 May Jun; 86(3):481-91. doi: 10.1111/j.1751-1097.2010.00735.x.
- M. E. Mottram and Harold A. Hill. Radiation Therapy of Ringworm of the Scalp. *Calif Med.* 1949 Mar;70(3):189-93. PMID: 18112454; PMCID: PMC1643734.
- Crossland PM. Therapy of tinea capitis; the value of x-ray epilation. *Calif Med.* 1956 May;84(5):351-3. PMID: 13316539; PMCID: PMC1531858.
- Wong CS, Strange RC, Lear JT. Basal cell carcinoma. *BMJ.* 2003 Oct 4;327(7418):794-8. doi: 10.1136/bmj.327.7418.794. PMID: 14525881; PMCID: PMC214105.
- Holíková Z, Massi D, Lotti T, Hercogová J. Insight into the pathogenesis of sporadic basal cell carcinoma. *Int J Dermatol.* 2004 Dec;43(12):865-9. doi: 10.1111/j.1365-4632.2004.02319.x. PMID: 15569004.
- Peris K, Fargnoli MC, Kaufmann R, Arenberger P, Bastholt L, Seguin NB et al. European consensus-based interdisciplinary guideline for diagnosis and treatment of basal cell carcinoma-update 2023. *Eur J Cancer.* 2023 Oct;192:113254. doi: 10.1016/j.ejca.2023.113254. Epub 2023 Jul 28. PMID: 37604067.
- Lichter MD, Karagas MR, Mott LA, Spencer SK, Stukel TA, Greenberg ER. Therapeutic ionizing radiation and the incidence of basal cell carcinoma and squamous cell carcinoma. The New Hampshire Skin Cancer Study Group. *Arch Dermatol.* 2000 Aug;136(8):1007-11. doi: 10.1001/archderm.136.8.1007. PMID: 10926736.
- Karagas MR, McDonald JA, Greenberg ER, Stukel TA, Weiss JE, Baron JA et al. Risk of basal cell and squamous cell skin cancers after ionizing radiation therapy. For The Skin Cancer Prevention Study Group. *J Natl Cancer Inst.* 1996 Dec 18;88(24):1848-53. doi: 10.1093/jnci/88.24.1848. PMID: 8961975.
- Albert RE, Omran AR, Brauer EW, Cohen NC, Schmidt H, Dove DC et al. Follow-up study of patients treated by x-ray epilation for tinea capitis. II. Results of clinical and laboratory examinations. *Arch Environ Health.* 1968 Dec;17(6):919-34. doi: 10.1080/00039896.1968.10665349. PMID: 5699300.
- Maalej M, Frikha H, Kochbati L, Bouaouina N, Sellami D, Benna F et al. Radio-induced malignancies of the scalp about 98 patients with 150 lesions and literature review. *Cancer Radiother.* 2004 Apr;8(2):81-7. doi: 10.1016/j.canrad.2003.12.003. PMID: 15063875.
- Shore RE, Albert RE, Reed M, Harley N, Pasternack BS. Skin cancer incidence among children irradiated for ringworm of the scalp. *Radiat Res.* 1984 Oct;100(1):192-204. PMID: 6494429.
- Boaventura P, Oliveira R, Pereira D, Soares P, Teixeira-Gomes J. Head and neck basal cell carcinoma prevalence in individuals submitted to childhood X-ray epilation for tinea capitis treatment. *Eur J Dermatol.* 2012 Mar-Apr;22(2):225-30. doi: 10.1684/ejd.2012.1670. PMID: 22381641.
- Shore RE, Moseson M, Xue X, Tse Y, Harley N, Pasternack BS. Skin cancer after X-ray treatment for scalp ringworm. *Radiat Res.* 2002 Apr;157(4):410-8. doi: 10.1667/0033-7587(2002)157[0410:scaxrt]2.0.co;2. PMID: 11893243.
- Mseddi M, Bouassida S, Marrekchi S, Khemakhem M, Gargouri N, Turki H et al. Carcinomes basocellulaires du cuir chevelu secondaires à une radiothérapie pour teigne: une série de 33 malades [Basal cell carcinoma of the scalp after radiation therapy for tinea capitis: 33 patients]. *Cancer Radiother.* 2004 Aug;8(4):270-3. French. doi: 10.1016/j.canrad.2004.03.006. PMID: 15450522.

15. Hassanpour SE, Kalantar-Hormozi A, Motamed S, Moosavizadeh SM, Shahverdiani R. Basal cell carcinoma of scalp in patients with history of childhood therapeutic radiation: a retrospective study and comparison to nonirradiated patients. *Ann Plast Surg.* 2006 Nov;57(5):509-12. doi: 10.1097/01.sap.0000229002.09605.5d. PMID: 17060730.
16. Shore RE, Moseson M, Harley N, Pasternack BS. Tumors and other diseases following childhood x-ray treatment for ringworm of the scalp (Tinea capitis). *Health Phys.* 2003 Oct;85(4):404-8. doi: 10.1097/00004032-200310000-00003. PMID: 13678280.
17. Poulalhon N, Dalle S, Balme B, Thomas L. Fast-growing cutaneous squamous cell carcinoma in a patient treated with vismodegib. *Dermatology.* 2015;230(2):101-4. doi: 10.1159/000368350. Epub 2015 Jan 24. PMID: 25633488.
18. Tauber G, Pavlovsky L, Fenig E, Hodak E. Vismodegib for radiation-induced multiple basal cell carcinomas (BCCs) of the scalp. *J Am Acad Dermatol.* 2015 Nov;73(5):799-801. doi: 10.1016/j.jaad.2015.07.016. Epub 2015 Aug 25. PMID: 26320385.

MULTIPLI BAZOCELULARNI KARCINOMI NAKON PRIMENE RADIOTERAPIJE U LEČENJU TINEA CAPITIS U DETINJSTVU: PRIKAZ SLUČAJA I PREGLED LITERATURE

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Sažetak

Uvod: Bazocelularni karcinom (BCK) je najčešći karcinom kože sa rastućom incidencijom širom sveta. Predilekciona mesta su glava i vrat u 80-85% slučajeva. Vodeći faktori rizika za nastanak ovog karcinoma su izlaganje UV zračenju i jonizujuće zračenje. Polovinom 20. veka, epilacija kapilicijuma izazvana zračenjem korišćena je kao efikasan metod u lečenju gljivičnih infekcija kapilicijuma.

Prikaz pacijenta: Predstavljamo slučaj osamdesetogodišnjeg muškarca sa šesnaestogodišnjom istorijom uza-stopne pojave multiplih BCK na kapilicijumu, licu i vratu. Zbog gljivične infekcije kapilicijuma u detinjstvu lečen je radioterapijom. Kliničkim i dermoskopskim pregledom uočeno je približno 25 BCK, pretežno na kapilicijumu. Najčešći tip BCK je bio nodularni, potom pigmentni i superficijalni tip. Histopatološki je potvrđena dijagnoza

BCK kod višestrukih lezija i jedan skvamocelularni karcinom (SCK). U ličnoj anamnezi kao najvažniji komorbidity izdvaja se leukemija vlasastih ćelija. Terapijski pristup zasnivao se na hirurškom lečenju i primeni 5-fluorouracil 5% krema.

Zaključak: Epilacija kapilicijuma izazvana zračenjem korišćena je kao efikasan metod za lečenje gljivičnih infekcija kapilicijuma. Jedan od neželjenih efekata bila je pojava BCK u područjima izloženim zračenju. Iako je hirurška ekscizija zlatni standard za lečenje BCK, primena 5% 5-fluorouracil krema može biti dobra opcija za pacijente sa multiplim BCK. Predstavljamo zadovoljavajuće rezultate nakon primenjene kombinovane terapije kod našeg pacijenta.

Ključne reči: bazocelularni karcinom, jonizujuće zračenje, radioterapija, tinea capitis, lečenje

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